

CASE REPORT

Giant Piloleiomyoma of the Forehead

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Cutaneous piloleiomyomas are benign smooth muscle tumors arising from the arrector pili muscles. Piloleiomyomas appear as firm dermal papules of skin color or with a reddish to brown surface, and are commonly located on the extremities. Histologically, these lesions are composed of interlacing bundles of smooth muscle cells in the reticular dermis. Our case presented with an unusually large nodule on the forehead that was accompanied by intermittent pain. Histological analysis was compatible with piloleiomyoma and the lesion showed haphazardly arranged bundles of smooth muscle in the dermis. We describe herein an interesting case of a giant piloleiomyoma occurring on the forehead. (*Ann Dermatol* 23(S2) S144~S146, 2011)

-Keyword-

Piloleiomyoma

INTRODUCTION

Leiomyomas are benign dermal tumors arising from the arrector pili muscles, the dartoic, vulvar, or mammary smooth muscles, or the muscles enveloping dermal blood vessels^{1,2}. They are subclassified into three groups according to the origin of the tumor^{1,2}: piloleiomyomas, genital leiomyomas, and angioleiomyomas. Among these, the

most common type is multiple piloleiomyomas^{1,3,4}. They vary in number and size and are often spontaneously painful or sensitive to touch and cold. The lesions can occur as grouped, linear, or dermatomal arrangements of firm, red to brown intradermal nodules³.

CASE REPORT

A 77-year-old man presented with a 5.5×4.5 cm erythematous nodule on his forehead. He said that the lesion began as an erythematous patch and progressively enlarged into a giant nodule over 50 years. Physical examination revealed a solitary indurated dermal nodule studded with yellowish papules and telangiectasia, which were prominent by dermoscopy and surface palpation (Fig. 1A). The patient often suffered from pain that was spontaneous or secondary to cold, pressure, or emotional disturbances.

Histopathological examination showed a hyperplastic epidermis, and the proliferation of sebaceous glands consisted of mature sebocytes. In addition, a relatively well circumscribed proliferation of smooth muscle cells filled the dermis and extended into the subcutaneous fat (Fig. 1B). The tumor cells usually exhibited elongated nuclei with blunt ends and formed multidirectional fascicles. Perinuclear clear zones were seen in the axial section of the tumor cells. No cellular atypia or mitotic activity was seen. Immunohistochemistry using Masson-Trichrome staining confirmed the interlacing bundles as smooth muscle fibers with interposing collagen bundles (Fig. 1C). The clinical and histopathological findings led to a diagnosis of a giant piloleiomyoma. We recommended that the patient undergo excision and skin grafting, but he did not want further treatment.

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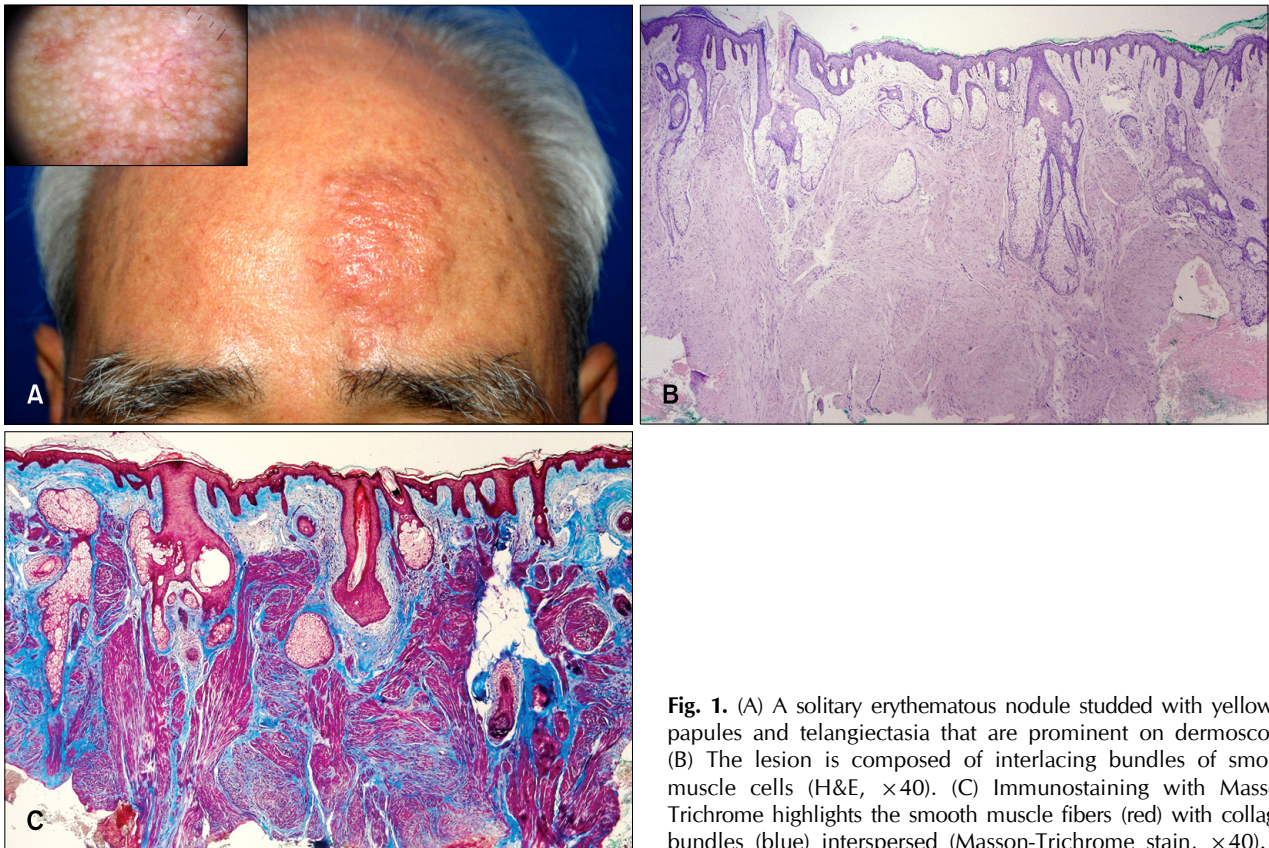


Fig. 1. (A) A solitary erythematous nodule studded with yellowish papules and telangiectasia that are prominent on dermoscopy. (B) The lesion is composed of interlacing bundles of smooth muscle cells (H&E, $\times 40$). (C) Immunostaining with Masson-Trichrome highlights the smooth muscle fibers (red) with collagen bundles (blue) interspersed (Masson-Trichrome stain, $\times 40$).

DISCUSSION

Cutaneous leiomyomas are uncommon benign smooth muscle neoplasms of the skin that are frequently unrecognized by clinicians³. The precise etiology of this disease is still unknown, but the involvement of a genetic predisposition has been suggested in reports on the familial occurrence of piloleiomyomas¹.

Piloleiomyomas are the most common type of cutaneous leiomyoma. They present as dermal, pink- or skin-colored papules or nodules, which are fixed to the skin. Piloleiomyomas are usually distributed in multiple lesions with each of them ranging from several millimeters to 1 cm in size. Their distribution is most commonly clustered, linear, or along Blaschko's line. The extensor extremities, trunk, and sides of the face and neck are the most common locations^{1,2}.

According to a study by Raj et al.⁴, which analyzed 53 cases of cutaneous piloleiomyomas in 45 patients, multiple piloleiomyomas are slightly more common than solitary lesions (ratio 1.2 : 1). There is an equal distribution between both sexes, and most patients are adults with a wide age distribution from 17 to 74 years. The limbs are a common site for both multiple and solitary

piloleiomyomas with solitary lesions favoring the limbs and multiple lesions the trunk. However, the forehead is a relatively uncommon site for a solitary piloleiomyoma, occurring in only 2 of 18 cases. In our case, the lesion was confined to the forehead as a solitary nodule. In addition, our case is interesting because the tumor was exceptionally large, at least 5 times larger than the largest previously documented piloleiomyoma. Raj et al.⁴ showed that the clinical size of multiple piloleiomyomas varied from a few millimeters to 1.5 cm with an average size of 0.7 cm. Moreover, the largest solitary lesion was 1.5 cm in diameter. Although Mitra et al.⁵ reported a case of a diffuse piloleiomyoma 15 cm in diameter on the right knee, there has been persistent controversy whether that case was a true piloleiomyoma because it is difficult to differentiate between smooth muscle hamartoma and leiomyoma. Other than this controversial case, to our knowledge, there are no reports of a cutaneous piloleiomyoma larger than the case we have presented.

In general, the tumors are painless initially but may become painful with time. The diagnosis may be facilitated by the characteristic provocation of pain on touching or chilling the lesion, although the cause for this remains unclear¹. Several authors believe the pain or

tenderness may be secondary to pressure on nerve fibers within the tumor, whereas others suggest it may be solely due to the contraction of muscle fibers³.

Subcutaneous nodules may be difficult to diagnose clinically because they do not have a characteristic surface change. However, it is possible to make a reasonable differential diagnosis if the nodule is painful. The acronym "ENGLAND" is often used to refer to these painful tumors: eccrine spiradenoma, neuroma, glomus tumor, leiomyoma, angioliipoma, neurilemmoma, and dermatofibroma⁶. With regard to the diagnosis of piloleiomyomas, the association with pain or a pseudo-Darier's sign due to muscle fiber contraction may be suggestive, and other painful tumors lack muscle on Masson trichrome stains^{1,3}. Histologically, piloleiomyomas are poorly circumscribed lesions consisting of interlacing smooth muscle fiber bundles with varying degrees of intermingled collagen³. They are located in the dermis and can infiltrate the surrounding tissue with extension into the subcutis¹. The smooth muscle fibers are fusiform and are composed of eosinophilic cytoplasm with elongated blunt-ended nuclei and perinuclear halos in cross-sections¹. In problematic cases, Masson-trichrome staining facilitates the differentiation of red smooth muscle fibers from the blue-green collagen. If necessary, desmin, smooth muscle actin, and muscle-specific actin may help to accentuate the smooth muscle origin of the tumor³.

Treatment of cutaneous leiomyomas depends on the degree of severity of symptoms and the extent of lesions. If only a few lesions are present, simple excision may be indicated^{1,3}. For those tumors with multiple lesions, medications such as nitroglycerin, nifedipine, and phenoxybenzamine have been used to treat symptomatic leiomyomas, but with limited success^{7,8}. In addition,

cryotherapy and electrocoagulation have shown little benefit⁹.

Our patient was unusual, especially in that he had a giant piloleiomyoma occurring on the forehead. A giant piloleiomyoma, although rare, should be considered in the differential diagnosis of an erythematous nodule with paroxysmal pain, especially on the face.

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